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CASE REPORT

Recurrent focal myositis in a patient on maintenance hemodialysis

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Summary Focal myositis is a benign inflammatory process involving a single group of muscles. It may resolve with conservative measures or may be a harbinger for polymyositis. Very few focal myosites are recurrent, and recurrence in patients with end-stage renal disease on hemodialysis is extremely rare. Clinical examination, electromyography, and magnetic resonance imaging help in identifying this entity. Muscle biopsy and histopathological evaluation are mandatory in diagnosis. It often responds to conservative treatment with anti-inflammatory drugs and physiotherapy, but occasionally it requires therapy with steroids.

局部肌炎是一種良性炎症，涉及單一肌肉組，可能會自行消失，亦可能是多發性肌炎的前身。局部肌炎的復發相當少見，在正在接受血液透析的末期腎病患者間更是極為罕見，其辨認一般透過臨床檢查、肌電學分析、及磁振造影，確診則必須透過肌肉活檢及病理組織檢驗達成。局部肌炎患者通常對物理治療及抗發炎藥物反應良好，僅在少數時候需要接受類固醇治療。

Introduction

Focal myositis is a benign inflammatory disease of a single group of muscles that presents as an isolated soft tissue mass. It is frequently localized in the lower limb and can be recurrent. Although there are a few reports of this entity in isolation, occurrence of the same in end-stage renal disease patients on hemodialysis is rare.[1] The exact etiology of this entity is not known. However, trauma, infection, and collagen vascular disease have been reported as causative factors. Often, it responds to nonsteroidal anti-inflammatory drugs (NSAIDs), or corticosteroids. Rarely, treatment with immunosuppressives, radiation, or surgical excision is warranted.

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Herein, we report a case of recurrent focal myositis in a patient with end-stage renal disease on maintenance hemodialysis.

Case report

A 45-year-old woman, diabetic and hypertensive, on maintenance hemodialysis, presented with sudden onset of painful swelling in the right calf. There was no history of fever, rash, or trauma. Her medications did not include statins, colchicine, or quinolones. There were no signs of fluid retention or cellulitis. The clinical suspicions were deep venous thrombosis and diabetic muscle infarction. Doppler ultrasound of the right lower limb venous system did not show evidence of thrombosis. Serum creatine phosphokinase (CPK) and lactate dehydrogenase were within normal limits. Ultrasound showed subcutaneous edema and increased echogenicity in the gastrocnemius and soleus muscles. Magnetic resonance imaging (MRI) showed altered signal intensity on T2W, T1W images in the gastrocnemius, soleus, and peroneus muscles (Fig. 1). Biopsy from the lateral head of the gastrocnemius showed few atrophic fibers, regenerating muscle fiber, and some fibers with hyalinization. The fascicles were separated by fibrous septae. There were areas of necrosis, crowding of nuclei, diffuse lymphoplasmacytic infiltration suggestive of focal myositis (Fig. 2). Electromyoneurography of upper and lower limb muscles was normal and thus the possibility of polymyositis was excluded. She improved with dextropropoxyphene, tramadol, and paracetamol.

One year later she presented again with sudden onset of a painful mass of 8 cm × 10 cm in the adductor compartment of the left thigh. The possibility of dialysis-related coagulopathy was ruled out. The bleeding time, clotting time, prothrombin time, activated partial thromboplastin time, and platelet count were within normal limits. Serum CPK was within normal limits. Ultrasound showed increased echogenicity in the adductor group of muscles. MRI of the thigh showed increased signal intensity on T2W images and decreased signal intensity on T1W in adductor magnus and semimembranosus, suggesting the possibility of focal myositis (Fig. 3). She improved with tramadol and paracetamol after 10 days.

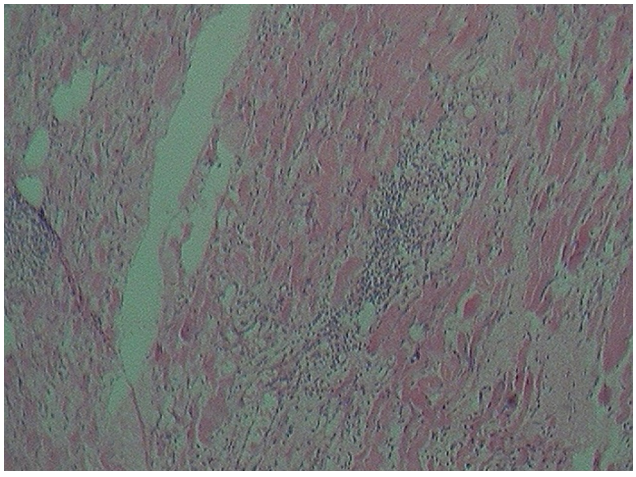

Figure 1 Hematoxylin and eosin stain showing degenerating and regenerating muscle fiber along with necrosis.

Figure 2 Magnetic resonance—short inversion time inversion recovery image showing increased signal intensity in the gastrocnemius and soleus muscles of the right leg.

Discussion

Focal myositis (FM) is a rare benign inflammatory disease of the skeletal muscles that often presents as a localized painful swelling. FM resolves spontaneously. Recurrence of this entity in different muscles on the ipsilateral or contalateral side have been described.[1] However, its recurring course in a patient on maintenance hemodialysis has not been described in the literature.

Sometimes, FM may be the heralding event of polymyositis.[2] Thrombophlebitis, myositis ossificans, nodular myositis, diabetic muscle infarction, amyloidosis, and proliferative myositis are a few differential diagnoses.[3] Diabetic muscle infarction is a close differential diagnosis but is associated with high CPK levels.

There are no specific laboratory tests to diagnose FM.[1] MRI often provides a sensitive means in diagnosing this entity, defining the extension and assessing disease activity and also in prognostication. The suggested findings on MRI are: enlargement of the involved muscle without invasion of adjacent structures, increased signal intensity on T2W images and decreased signal intensity on T1W image.

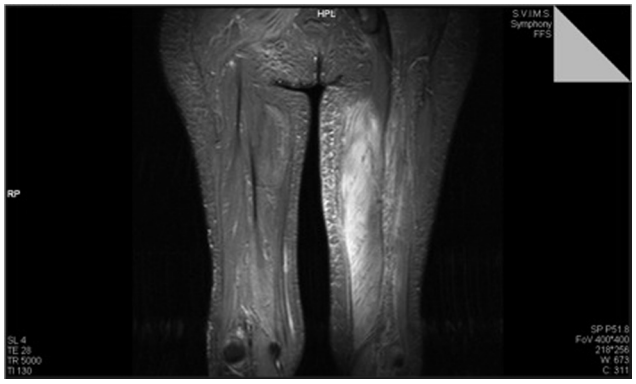

Figure 3 Magnetic resonance—short inversion time inversion recovery image showing increased signal intensity in the left adductor magnus and semimembranosus muscles.

Electromyography (EMG) may reveal short duration motor unit potential coupled with increased polyphasia and normal EMG in the other muscles.[2]

Etiopathogenesis is not well described in the literature. The role of proliferation of interstial connective tissue, trauma, and underlying infection are a few of the plausible causes. Clinical evaluation and imaging may not differentiate malignant soft tissue tumor and FM. Hence muscle biopsy is mandatory and histopathological examination is confirmatory. This shows densely packed muscle fibers arranged in fascicles or rounded clusters enveloped by bundles of connective tissue. There is marked variation of fiber diameter, increased number of internalized nuclei, fiber splitting, and occasional degenerating and regenerating muscle fibers. There is also severe endomysial and perimysial fibrosis with characteristic pattern of large nests of muscle fibers forming lobules. The features that help in distinguishing FM from polymyositis are the large size of muscle fiber nests forming tightly packed nodules, enveloped by fibrosis, with exaggerated fiber hypertrophy and mostly T cell and macrophage infiltrate. Immunohistochemistry reveals scant T cells, with few B cells and macrophages in the degenerating muscle fibres.[2] In FM, there is never an involvement of lymphocytic infiltration in vital muscle fibers as is seen in polymyositis.

In the majority of cases, the evolution of FM is self-limited and responds to conservative management. Surgical excision, radiation therapy, immunosuppressive agents, NSAIDs are the treatment strategies. Relapses are not rare. Normal muscle enzyme level, absence of inflammatory markers (ESR/C-reactive protein) suggests a benign outcome. Elevated CPK or ESR suggests evaluation for polymyositis.[1]

In our patient, FM was recurrent and responded to dextropropoxyphene, tramadol, and paracetamol. FM is always a diagnosis of exclusion and is confirmed by histology.

In most patients, FM improves over a period of time with NSAIDs (COX-2 inhibitors). If ESR or serum CPK are abnormal, EMG suggest involvement of other muscles and muscle biopsy is not consistent with FM, early treatment with steroids should be considered.

The points of interest in our presentation are: (1) common presentation of a less common entity; and (2) recurrence in contralateral limb muscles.

Conflicts of interest

All authors declare no conflicts of interest.

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